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Leiomyomatosis Peritonealis Disseminata: A Case Report

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Abstract **Case Report**

***Introduction*:** Leiomyomatosis peritonealis disseminata (LPD) manifests as a distinctive benign clinical entity marked by the proliferation of nodules within the peritoneal and subperitoneal layers. LPD poses diagnostic challenges due to its unique presentation, yet it typically follows a benign trajectory with minimal risk of malignant transformation. ***Presentation of Case*:** A 38-year-old female with a pain in the left iliac fossa which has progressed to a gradual distension of the abdomen, proceeded to our institution for neoadjuvant treatment of abdominal and pelvic masses which were firstly considered as desmoid tumor. The histologic diagnosis of the mass lesions revealed smooth muscle benign cells. This is the first case of LPD reported in Casablanca Oncology and Radiotherapy Ibn Rochd Center. A meticulous review of the literature was conducted as well. ***Discussion*:** The differential diagnosis of LPD is difficult due to its clinical resemblance with intra-abdominal desmoid tumor, peritoneal carcinomatosis, metastatic lesions and with benign metastasizing leiomyoma as well. Etiological factors, pathophysiology and clinical manifestations which lead to a safe diagnosis of LPD are adequately described. ***Conclusion*:** Leiomyomatosis peritonealis disseminata (LPD) is a rare benign condition prone to recurrence, particularly in patients with a history of uterine fibroid surgery. Biopsy is essential for differential diagnosis. We recommend, based on the experience from this clinical case, that an exploratory laparotomy should be performed whenever the clinical and histological context raises suspicion of LPD, to offer a better chance of effective management. For patients who wish to preserve their fertility, medical treatment with aromatase inhibitors or selective progesterone receptor modulators may facilitate conservative surgery options. For patients without reproductive desires, total abdominal hysterectomy, bilateral salpingo-oophorectomy, and debulking of affected tissue are recommended. However, involvement of neighboring organs or anatomical structures may suggest non-resectability. Regular follow-up with imaging and clinical assessment is essential to monitor for recurrence or malignant transformation of LPD, ensuring timely intervention and better outcomes.

Keywords: Case Report, Leiomyomatosis Peritonealis Disseminata, Laparoscopic Myomectomy.

1. INTRODUCTION

Leiomyomatosis peritonealis disseminata (LPD) is characterized by dissemination and proliferation of peritoneal and subperitoneal lesions primarily originated from smooth muscle cells. LPD is more common in premenopausal women. Although benign in nature, LPD may degenerate into peritoneal leiomyosarcoma [1,2].

So far, no more than 200 cases of LPD have been reported. However, the published case reports concerning LPD following laparoscopic myomectomy are increasing. Preoperative diagnosis of LPD is presented as a challenge due to unspecific clinical manifestations and its clinical resemblance with peritoneal carcinomatosis or metastatic lesions.

Herein, we reported 1 case of LPD from the Oncology and Radiotherapy VI Center in Ibn Rochd University Hospital in Casablanca, March 2024.

2. CASE REPORT

A 38-year-old female proceeded to our institution for neoadjuvant treatment of abdominal and pelvic masses. Approximately one year ago, she presented with left iliac fossa pain, which progressed to a gradual distension of the abdomen and abdominal discomfort. This prompted the patient to initially seek consultation at the surgical department. The Initial pelvic MRI that had been initially performed had revealed a an inter-uterorectal pelvic mass measuring 116*94 mm, exhibiting solid-cystic heterogeneous characteristics, extending over 143 mm. It intimately contacts the left psoas major muscle head and intimately abuts the L5-S1 intervertebral disc with loss of the disc margin. It also contacts the rectum with loss of the fat plane separation, laterally, it contacts the left external and internal common iliac veins bilaterally. It also contacts the body and cervix of the uterus with loss of the separating margin. A right ovarian mass with similar characteristics to the above-described mass measures 97.4*71 mm, extending over 117 mm. She does not have a history birth neither of laparoscopic myomectomy, neither previous history of prolonged use of contraceptive pills. Furthermore, she had family history of nor leiomyomas, neither leiomyosarcomas of the uterus. The first diagnosis was desmoid tumor. In order to exclude the probability of malignancy, Fine needle aspiration biopsy was recommended.

Histological analysis of the tissue sample revealed initially a desmoid tumor. Another biopsy had been released to confirm the diagnostic. Morphological appearance was compatible with disseminated peritoneal leiomyomatosis.

Providing the patient's findings and clinical history she was referred to our institution for further evaluation.

Angio-CT scan was performed 02/2024 (figure 1) to analyse vascular proximity, it showed a large solid-cystic intra-peritoneal mass with well-defined polylobed contours, exhibiting heterogeneous enhancement of the fleshy portion following contrast injection, measuring 21.2*16.4 cm, extending over 19 cm (compared to 11*10 cm extending over 14 cm). There was Intra and peri-tumoral neoangiogenesis and significant intra- and retroperitoneal effusion. Left ureteral compression with moderate upstream uretero-pyelocaliceal dilatation, closely associated with the descending aorta and right renal artery. It displaces the celiac trunk and superior and inferior mesenteric arteries to the left. It compresses the primitive iliac arteries, external and internal. It compresses the portal trunk and spleno-mesenteric confluence. A CT scan of the chest showed no abnormalities. The tumor markers CA19-9, ACE, CA15-3, and CA125 were negative.

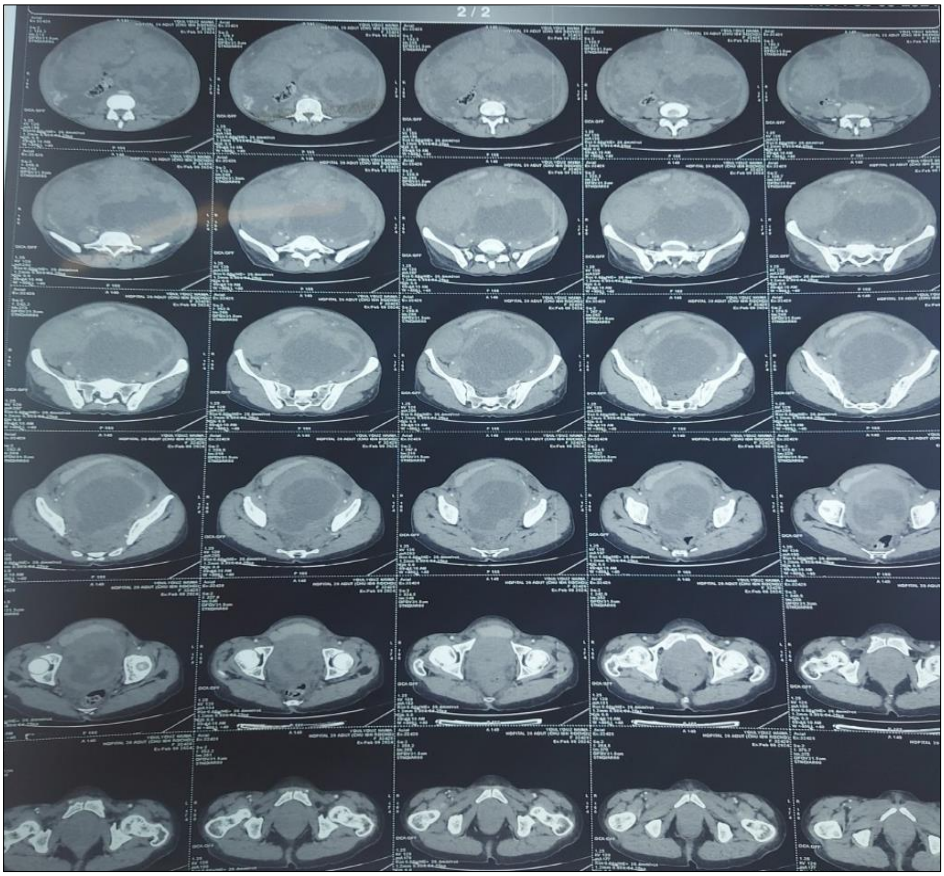

Fig. 1: CT abdominal scan showing multiple tumescent masses.

The physical examination of the patient was unremarkable. The vital signs and blood test results were all into the normal spectrum. The case file was presented at the multidisciplinary team meeting (RCP). An exploratory laparotomy was scheduled to explore the involvement and contact with surrounding vessels and adjacent organs, ensuring a thorough assessment of any potential invasion or non-resectability. During the operation on 03/2024, the oncology surgeons were surprised to find that the masses were encapsulated and not in contact with surrounding vessels or adjacent organs, despite MRI findings indicating contact with the vessels, consequently, The hystero-salpingo-oophorectomy and pelvic lesions resection (figure 2) were performed ,given that the patient did not wish to conceive. The histopathologic analysis of the tissue sample from the excisional nodules revealed LPDs(figure 3 A and). No recurrence was found during the postoperative 7 months follow-up.

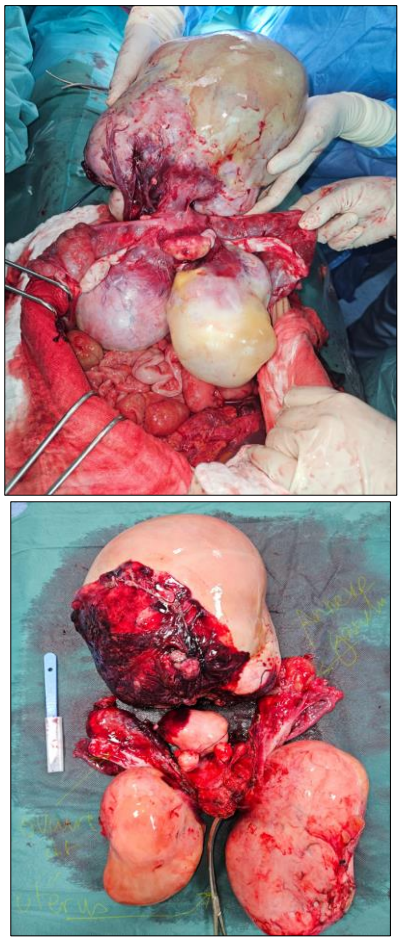

Figure 2 : Hystero-salpingo-oophorectomy and pelvic lesions resection

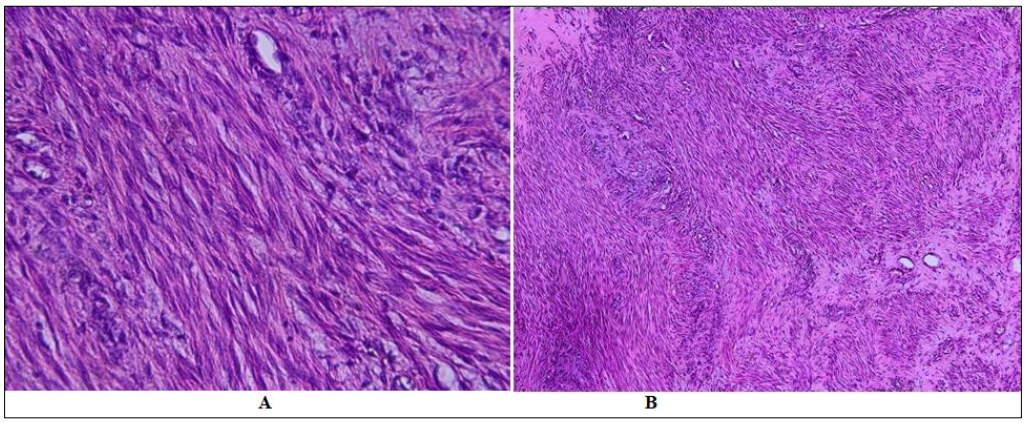

Figure 3: A: Tumor proliferation arranged in long interwoven bundles (Hematoxylin-eosin, x100). B: pindle-shaped cells with elongated nuclei, without atypia (Hematoxylin-eosin, x400)

3-DISCUSSION

LPD, first described by Wilson *et al*., in 1952, is an extremely rare yet typically benign condition resulting from uterine fibroids or the dissemination and implantation of tissue onto the surfaces of the greater omentum, mesentery, and colorectum, also referred to as parasitic leiomyoma [3].

Presently, fewer than 200 cases of LPD have been revealed worldwide. Its etiology and pathophysiology remain unclear. It has been proposed that LPD could be caused by metaplasia of mesenchymal cells of the peritoneum and, in susceptible women, residual myoma in the abdominal cavity might contribute to the development of LPD [4]. Metaplasia and differentiation from mesenchymal stem cells into smooth muscle cells may be promoted by estrogen exposure [5].

Therefore, LPD is commonly regarded as a benign condition occurring predominantly in premenopausal women. However, there have been rare instances of LPD diagnosed in menopausal women or cases where malignant transformation has occurred [6,7].

Iatrogenic origin subsequent to laparoscopic surgery has been suggested as another main theory of the etiology and pathophysiology of LPD in setting of development and widely application of laparoscopic uterine fibroids morcellation [8].

In 1997, Ostrzenski [9], reported the first case of uterine leiomyoma particles growing in an abdominal-wall incision after laparoscopic retrieval. Since then, there has been a growing number of reports on iatrogenic LPD following laparoscopic surgery in recent decades. In this study, the patient did not undergo laparoscopic uterine fibroid morcellation. Therefore, laparoscopic uterine fibroid morcellation may not be the primary factor for the secondary dissemination of fibroids.

The preoperative diagnosis of LPD poses a challenge as patients often present with asymptomatic or atypical symptoms such as abdominal pain, discomfort, and irregular vaginal bleeding. Erenel *et al*., [10], described 53 patients with LPD, among whom 28 (53%) cases presented with abdominal pain, 13 were asymptomatic, and others had a palpable pelvic mass. In this study, the patient presented with gradual distension of the abdomen, abdominal discomfort and abdominal masses. The diagnosis of LPD in this case was confirmed through preoperative biopsy and pathological evaluation. It is important to discuss the concerning relationship between the tumor and nearby blood vessels, which may lead surgeons to reconsider surgical exploration.

Pathologically, PDL peritoneal disseminated Leiomyoma is characterized by its multifocal nature, with nodules consisting of fusiform smooth muscle cells and occasional fibroblasts, lacking atypia or mitosis. The morphology resembles typical uterine leiomyoma with a vortex pattern, though PDL can occasionally appear as different cell types. Differential diagnoses for DPL include; GIST: the most common mesenchymal tumor of the digestive tract, characterized by spindle and epithelioid cells, with CD117, CD34, and Dog-1 expression [11].

Primary Intraperitoneal Leiomyosarcoma:

This sarcoma usually originates from the retroperitoneum, presenting with atypical cells and high mitotic activity, distinguishing it from DPL [12].

Desmoid Tumors:

These are benign fibrous tumors that can occur in the abdominal cavity and may mimic DPL radiologically. They are characterized by aggressive local growth but do not metastasize [13].

Ovarian Tumors:

Tumors such as ovarian leiomyomas or other neoplasms can present as pelvic masses and should be considered in the differential diagnosis, especially when imaging shows adnexal involvement [14].

Angiomyolipoma:

A benign tumor comprising vascular, adipose, and smooth muscle tissues, identifiable through immunohistochemical markers [15].

Multiple Abdominal Leiomyomas:

Although pathomorphologically similar to DPL, they can be differentiated through medical, hormonal, and genetic histor [16]. In our case report, a desmoid tumor was initially suspected based on radiological findings and the initial pathological assessment. However, after a second biopsy, the diagnosis of disseminated peritoneal leiomyomatosis (PDL) was confirmed.

Although LPD is benign, its imaging characteristics of pelvic and abdominal dissemination mimic malignancy. Therefore, when combined with elevated serum CA125 levels and disseminated implantation, distinguishing LPD from malignancy becomes challenging. The preoperative diagnosis of LPD relies on a comprehensive evaluation of medical history and imaging studies, particularly in patients with a history of laparoscopic uterine fibroid morcellation. While ultrasonography, CT, and MRI are effective diagnostic tools, they offer limited assistance in differentiating malignancies. Ultimately, histopathologic examination of preoperative biopsy tissues is essential to confirm the diagnosis of LPD. It is also crucial to emphasize the necessity of exploratory laparotomy for accurate staging and surgical intervention. This approach allows for a comprehensive assessment of the tumor's extent and facilitates appropriate treatment planning when histopathological diagnosis is not made.

Very few cases of LPD undergo malignant transformation [17,18], with only about 10 reported cases to date. Risk factors for malignant transformation of LPD include the absence of a history of oral contraceptives, pregnancy, or uterine leiomyomas, as well as the lack of expression of estrogen and progesterone receptors in leiomyoma nodules, and recurrence within 1 year after initial treatment. For patients with high-risk factors, regular follow-up should include vaginal ultrasound and pelvic and or abdominal MRI examinations. Surveillance for LPD typically occurs every 3 to 6 months during the first two years, then shifts to annual evaluations based on individual risk factors and findings. Regular imaging is vital for the early detection of any recurrence or disease progression, which may indicate a malignant form [19].

Currently, there are no established treatment guidelines for LPD due to its rarity. This absence of clear protocols challenges management and necessitates individualized treatment strategies. Recently, it has been suggested that surgical therapy should be personalized based on factors such as the patient's age, symptoms, desire for childbearing, and past treatments. In principle, surgical intervention should aim to remove all tumor nodules to the greatest extent possible [20].

For women who do not desire reproduction, a more aggressive surgical approach may be considered, including total abdominal hysterectomy, salpingo-oophorectomy, and debulking. However, the question of whether fibroids on the surface of the intestinal wall should be primarily resected remains debatable. Considering this patient without child-bearing requirements, only hysterectomy and salpingo-oophorectomy were performed with resection of fibroid nodules. No recurrence was observed during the 7-month postoperative follow-up. Additionally, patients suspected of LPD should be informed prior to surgery that extensive and complicated procedures may be necessary. Furthermore, susceptible dissemination sites of LPD nodules should be carefully explored to minimize the risk of postoperative recurrence.

LPD is closely linked with estrogen, leading to suggestions that gonadotropin-releasing hormone injections, aromatase inhibitors, or selective progesterone receptor modulators could be used for conservative or primary treatment in young women with reproductive desires, as well as for preventing postoperative recurrence [21,22]. However, the efficacy and safety of these drugs in LPD patients require confirmation through further clinical case studies, given the current lack of clinical evidence.

4-CONCLUSION

Leiomyomatosis peritonealis disseminata (LPD) is a rare benign condition with a risk of recurrence, especially in patients with a history of uterine surgery for fibroids. A biopsy is crucial for establishing a differential diagnosis.

e recommend, based on the experience from this clinical case, that an exploratory laparotomy should be performed whenever the clinical and histological context raises suspicion of LPD, to offer a better chance of effective management. For patients who wish to preserve their fertility, medical treatment with aromatase inhibitors or selective progesterone receptor modulators may facilitate conservative surgery options. For patients without reproductive desires, total abdominal hysterectomy, bilateral salpingo-oophorectomy, and debulking of affected tissue are recommended. However, involvement of neighboring organs or anatomical structures may suggest non-resectability.

Regular follow-up with imaging and clinical assessment is essential to monitor for recurrence or malignant transformation of LPD, ensuring timely intervention and better outcomes.

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